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Published in:
Developmental Review

DOI:
[10.1016/j.dr.2020.100913](https://doi.org/10.1016/j.dr.2020.100913)

Publication date:
2020

Document Version
Publisher's PDF, also known as Version of record

[Link to publication in Tilburg University Research Portal](#)

Citation for published version (APA):

De Caluwé, E., Vergauwe, J., Decuyper, M., Bogaerts, S., Rettew, D. C., & De Clercq, B. (2020). The relation between normative rituals/routines and obsessive-compulsive symptoms at a young age: A systematic review. *Developmental Review*, 56, [100913]. <https://doi.org/10.1016/j.dr.2020.100913>

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The relation between normative rituals/routines and obsessive-compulsive symptoms at a young age: A systematic review

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ARTICLE INFO

Keywords:

Ritual
Obsessive-compulsive disorder
Child
Continuity hypothesis
Integrative conceptual model

ABSTRACT

Objective: One of the most challenging issues in the pediatric obsessive-compulsive (OC) disorder field is to differentiate pathological OC symptoms from the phenotypically similar normative rituals/routines that characterize normal childhood development. Given their similarities, it can be questioned whether both constructs reflect two qualitatively distinct behavioral expressions of different etiological factors or rather reflect two diverse manifestations of one single continuum (cfr. the continuity hypothesis).

Method: This paper aims to improve our understanding of the relation between normative childhood rituals/routines and OC symptoms in two ways. First, the continuity hypothesis was investigated by systematically reviewing current evidence on this association, using various databases (Web of Science Core Collection, MEDLINE/PubMed, and ScieLO Citation Index) from 1950 until February 1, 2019 (registration number: CRD42019121293). Second, based on this systematic review, an integrative conceptual model was developed describing this relation from different perspectives on sources of influence.

Results: The literature search initially revealed 2122 hits, with 114 full-texts being assessed for eligibility. After applying several selection criteria, 18 studies were included in the review. The results generally support the continuity hypothesis and reveal important etiological factors at different levels of behavioral analysis, including the phenotypic (anxiety), neurobiological and genetic level. Also age and the presence of other disorders appeared to be important factors in evaluating the level of normality.

Conclusion: We provide a conceptual framework to inform future research aimed at improving the understanding of the relationship between normative rituals/routines and pathological OC symptoms. Conceptual implications are discussed and clinical recommendations are given to improve early identification and differentiation.

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<https://doi.org/10.1016/j.dr.2020.100913>

Received 15 August 2019; Received in revised form 12 April 2020

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Introduction

Obsessive-Compulsive Disorder (OCD) is characterized by uncontrollable, intrusive and time-consuming thoughts or images (obsessions) and/or repetitive acts or mental rituals (compulsions) that interfere with daily functioning (American Psychiatric Association [APA], 2013). The content of these obsessions and compulsions generally involves themes of contamination, washing, aggression, symmetry, ordering, repeating and checking (Leckman, Zhang, Alsobrook, & Pauls, 2001; McKay et al., 2006). OCD was historically considered a rare condition in youth (Geller, 2006), until studies indicated that pediatric prevalence rates were considerably higher than initially assumed (Leonard, Ale, Freeman, Garcia, & Ng, 2005; Merlo & Storch, 2006). Currently, OCD is one of the more common pediatric psychiatric disorders. It affects 1–3% of the children and adolescents (Heyman et al., 2001; Thomsen, 2013) with an average 3:2 male-to-female ratio (Geller et al., 2012). Moreover, approximately half of the adults suffering from obsessive-compulsive (OC) symptoms report an onset prior to adulthood (Stewart et al., 2004), typically between the ages of 6 and 14 (Garcia et al., 2009; Geller et al., 1998). Beyond these retrospective ratings that might be affected by recall bias (Leonard, Goldberger, Rapoport, Cheslow, & Swedo, 1990), prospective data indicate that some OC symptoms observed in children as young as three years old (Garcia et al., 2009) have predictive validity in terms of OC-related outcomes (Geller et al., 2012), underscoring the clinical importance of early identification.

One of the challenging issues in the early identification of pediatric OCD, however, is that typically developing children may also show normative behaviors that are phenotypically similar to those associated with OCD (Evans & Maliken, 2011; Evans et al., 1997; Zohar & Bruno, 1997). These typical behaviors consist of normative rituals and routines, which are mostly used interchangeably (Fiese et al., 2002) and reflect a broad range of ritualistic, routinized, repetitive, compulsive-like and tic-related behaviors. For example, beginning around the age of two, children may insist on sameness in daily routines, pay attention to minute details such as imperfections in toys and clothes (it has to be/feel 'just right') and display mealtime rituals (needing to have the food presented in a certain way). Whereas parents may install a bedtime routine (e.g., bath, book reading and kissing goodnight), the child may begin to ritualize it by focusing on specific details or specific manners in which the routine is installed (Evans & Maliken, 2011; Evans et al., 1997; Gesell, Ames, & Ilg, 1974). Similar to pathological compulsions, interruptions in these typical rituals/routines may result in distress and the child insisting to start all over again (Evans, Elliott, & Packard, 2001).

From a normative developmental perspective, it has been argued that these rituals/routines provide order and predictability for young children who have little understanding of the contingencies in daily life (Piaget, 1950). Hence, these rituals/routines are common and considered normative and even adaptive in early childhood (Evans & Maliken, 2011). In the context of OCD, however, pathological variants of these rituals/routines (i.e., OC symptoms) adversely affect multiple aspects of functioning and are therefore important targets for early clinical intervention. Toward this end, distinguishing clinically significant behavior from normative behavior is an important objective for clinicians, and requires a more firm understanding of the relation between these phenotypically similar constructs.

Distinguishing normative rituals/routines versus clinical OC symptoms in youth

The main question to be answered in this respect is the extent to which normative rituals/routines and pathological OC symptoms are qualitatively distinct behavioral expressions with different etiological factors versus diverse manifestations along a single continuum. This question can be addressed from the continuity hypothesis, stating that the relationship between normality and psychopathology is primarily one of severity or degree, with both kinds of behaviors reflecting similar underlying factors (Krueger & Tackett, 2003; Rettew, 2013; Tackett, 2006; Werner, 1948, 1957). Although increasing evidence indeed suggests that the nature of psychopathology is fundamentally dimensional (Krueger et al., 2018), only a limited number of studies explicitly focused on the nature of the relation between normative rituals/routines and maladaptive OC symptoms (i.e., continuity or not?). In addition, potential influencing factors of this relationship (i.e., both explanatory/etiological and moderating factors) have not been comprehensively addressed, and may contribute our understanding in this regard.

Moreover, besides the fact that early ritualistic behaviors occur during both normative childhood development and pediatric OCD, they are also displayed in the context of other disorders, such as autism spectrum disorders, tic disorders, and intellectual disabilities (Evans, Uljarevic, Lusk, Loth, & Frazier, 2017; Geller et al., 2012; Geller, 2006). Such widespread presence of rituals/routines across disorders makes it even more important to evaluate their degree of normality/pathology, specifically addressing an evaluation of the level of impairment for the child and his or her social network. From a treatment perspective in particular, it is useful to additionally examine to what extent the rituals/routines occur in the context of (coping with) other vulnerabilities, as different etiological liabilities may need a different treatment approach (e.g., CBT vs SSRI) (Geller et al., 2012; Geller, 2006). Hence, from this intervention perspective, all established disorders in which rituals/routines may occur are important to consider as well, because they may guide the clinician toward the most appropriate intervention of pathological rituals/routines in the context of the broader clinical picture of the child.

The current study

The primary objective of this study is to investigate the continuity hypothesis by systematically reviewing all literature on the relation between normative rituals/routines and OC symptoms in children and adolescents. Based on this review, an integrative conceptual model that may serve as a guiding framework for both researchers and clinicians will be proposed as a secondary objective. This conceptual model will integrate important factors that might be systematically considered in evaluating the

maladaptive character of the presented rituals/routines. In addition, in order to guide clinicians toward a tailored intervention of maladaptive ritualistic behaviors, all disorders/conditions that can include rituals/routines will be summarized in this model. Hence, the current paper aims to improve our understanding of the *nature* of the relationship between rituals/routines and OC symptoms, taking into account potential *influencing factors* of this association and addressing the broader clinical picture (i.e., *other disorders*) in which routines/rituals may occur. In doing so, this paper will offer various suggestions for further research and recommendations to improve a valid identification of early OC symptoms and differentiation from normative expressions, hence contributing the field in the search for evidence-based assessment and treatment decisions. This early focus is of crucial importance because an early onset of OC symptoms is one of the strongest predictors of a negative prognosis over time (Stewart et al., 2004) and the earlier the identification and treatment, the better the outcome (Walitza et al., 2011).

Method

Systematic review

The continuity hypothesis was investigated by conducting a systematic review according to the PRISMA guidelines (Liberati et al., 2009; Moher, Liberati, Tetzlaff, Altman, & Group, 2009). To reduce publication bias and provide greater transparency, the review protocol is registered in PROSPERO (CRD42019121293). To identify studies on the relation between normative rituals/routines and OC symptoms in children and/or adolescents, we performed a systematic search in various Web of Science databases (including Web of Science Core Collection, MEDLINE/PubMed, and SciELO Citation Index) from 1950 until February 1, 2019. This search was constrained to English peer-reviewed articles. We focused on studies with at least one search term in the title for each of the following aspects:

- I. Normative rituals/routines: *ritual** OR *rouin** OR *repet** OR *tic** OR *norma** OR *develop**
- II. OC symptoms: *obsessi** OR *compuls** OR *repet** OR *OCD**
- III. Children and/or adolescents: *child** OR *toddler** OR *adolescen** OR *youth** OR *young** OR *teen** OR “*primary school*” OR “*middle school*” OR “*high school*” OR “*secondary school*” OR “*young people*” OR *pediatric** OR *early*

To guarantee that all relevant articles were included, we did a second search using I and II, but omitting III because age groups are not always mentioned in titles. Also ‘*repet**’ was excluded in II, as it was already included in I (to capture all relevant articles). We also added ‘NOT #1’ to exclude articles from the first search. The second author reviewed the search strategies and disagreements were resolved by consensus.

The first author screened all articles based on title (and abstract). Irrelevant articles were excluded, i.e., if they: 1) did not focus on normative rituals/routines or OC symptoms, or 2) did not address younger age groups. To test objectivity and inter-rater agreement, the second author independently screened 10% of the articles based on title (and abstract). Then, the first author reviewed the full-texts of the selected articles on eligibility criteria. These were: 1) the inclusion of both key constructs in the paper (i.e., normative rituals/routines and pathological OC symptoms), and 2) participants being children and/or adolescents (≤ 18 years old). Besides the database search, reference lists of selected studies were checked (backward citation search). Studies (with varying designs) were included if they focused on both normative rituals/routines and OC symptoms in children and/or adolescents. Participants included typically developing children/adolescents and/or pediatric OCD patients. Both key constructs could be investigated in the same sample, or studies were also included if normative rituals/routines were investigated in a typical sample and OC symptoms in an OCD sample. Studies were excluded if they: 1) focused on one key construct only (i.e., only normative rituals/routines; or only pathological OC symptoms), 2) were exclusively conducted in other patient groups (e.g., with Down syndrome), 3) were exclusively conducted in adults, 4) were not peer-reviewed articles (e.g., meeting abstracts), 5) were not conducted in humans, or 6) had no English full-text. To prevent reprint bias, the results of the selected articles were carefully investigated and their variables were extracted into a table, including: first author, publication year, main conclusion, (contra) evidence for continuity (reflecting the main variable of interest), key constructs, sample size, participant characteristics (without any reports of comorbidity), participants’ age, and study design characteristics.

Development of an integrative conceptual model

Based on the systematic review, we propose a conceptual model that (1) describes the relation between normative rituals/routines and OC symptoms at a young age, and (2) integrates factors that may systematically influence (i.e., explain or moderate) this relation, such as developmental aspects (e.g., age) as well as other disorders that complicate the understanding of ritualistic behaviors from a diagnostic point of view, because they can all contain rituals/routines in their symptomatic picture (e.g., autism spectrum disorders, tic disorders, intellectual disabilities). Towards this end, a comprehensive list of all disorders that can be characterized by rituals/routines is integrated in this model, which may help clinicians to evaluate ritualistic behaviors that are considered as maladaptive in the context of the most suitable underlying diagnosis. These disorders were not included in the systematic search, however, as this would result in too many irrelevant hits for the scope of the current review, which primarily focuses on the relation between normative rituals/routines and OC symptoms. Therefore, we do also not provide a discussion on all these disorders and on how they relate to rituals and OCD. Nevertheless, by screening all hits from the search, we were able to document the full spectrum of disorders that may comprise rituals/routines.

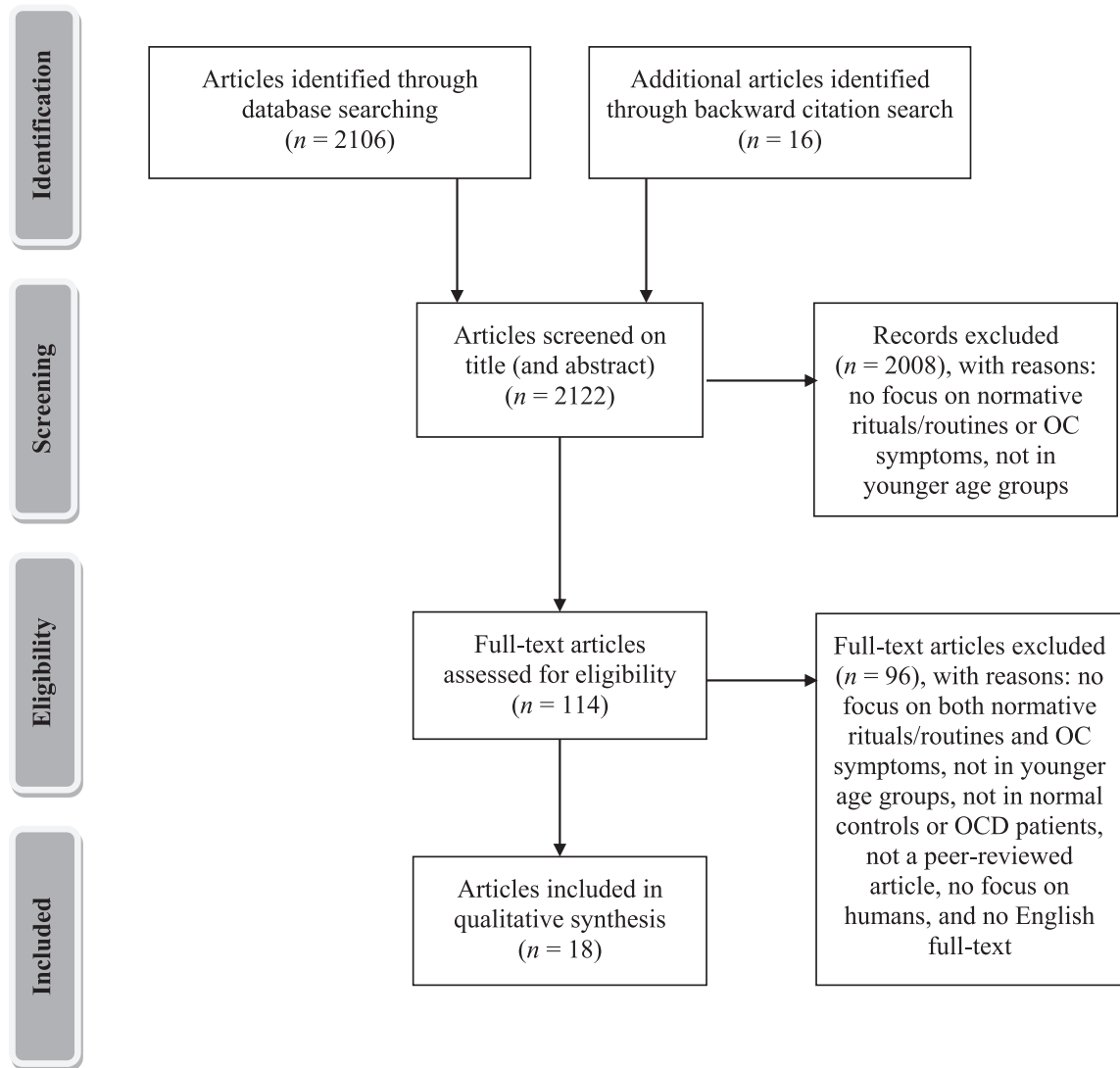


Fig. 1. PRISMA flow diagram of study selection.

Results

Systematic review

The database search resulted in 2106 hits (1261 from Search 1 and 845 from Search 2). The backward search revealed 16 hits, together resulting in 2122 unique articles. The first author screened all 2122 titles (and abstracts) and excluded them if they: 1) did not focus on normative rituals/routines or OC symptoms, or 2) did not address younger age groups. The second author did the same for 10% of the hits and showed 99% inter-rater agreement with the first author, reflecting a very good $\kappa = 0.86$ (95% CI, 0.57–1), $p < .001$. Subsequently, the first author carefully assessed the full-texts of the remaining 114 articles for eligibility, of which 18 were included in the review (Fig. 1). Based on these studies (Table 1), evidence for the continuity hypothesis is discussed, largely organizing the results of the studies along the following perspectives: 1) clinical/phenotypical (targeting exclusively the association between normative rituals/routines and OC symptoms, or adopting anxiety as additional construct), 2) developmental, 3) neurological, and 4) behavioral-genetic perspectives (note that several studies combine perspectives).

Normative rituals/routines and obsessive-compulsive symptoms on a continuum

In their pioneering study, Leonard et al. (1990) investigated the continuity between childhood OC symptoms and normative rituals/routines by comparing 38 OCD-diagnosed children and adolescents (7–18 years old) with 22 controls (9–18 years old) on early rituals and superstitions. They hypothesized that OCD is not on a continuum with superstitions and normative rituals, due to

Table 1
Evidence and Contra Evidence for Continuity between Normative Rituals/Routines and Obsessive-Compulsive (OC) Symptoms.

Authors, year (Chronological)	Main conclusion	Evidence	Contra evidence	for continuity between:	N	Participants (+age)	Study design
Flament et al. (1988)	no difference in intensity or number of superstitions/rituals	X		normal and abnormal obsessions; rituals and compulsions	18 + 338	adolescents with OCD + controls	epidemiologic interview study
Swedo et al. (1989)	no difference in intensity or number of superstitions/rituals	X		normal and abnormal obsessions; rituals and compulsions	70	children & adolescents with OCD	review study
Leonard et al. (1990)	no difference in intensity or number or type of superstitions (and rituals, but no clear answer)	X	X	normal and abnormal obsessions; rituals and compulsions	38 + 22	children & adolescents with OCD (7–18 years old) + controls (9–18 years old)	cross-sectional interview study
Zohar and Bruno (1997)	the same rituals are common and adaptive at younger age but rare and maladaptive at older age (related to anxiety)	X		rituals and compulsions	1083	community sample of children & adolescents (8–14 years)	cross-sectional questionnaire study
Evans et al. (1999)	parallels are drawn between “normative compulsions and fears” and “compulsions and anxieties found in OCD”; fears-rituals relation increases with age and becomes maladaptive	X		normal fears and abnormal anxieties; rituals and compulsions	61	typically developing children (1–7 years old)	cross-sectional questionnaire study
Evans et al. (2001)	common underlying visuospatial abilities	X		rituals and compulsions	51	typically developing children (3–13 years old)	neurological tasks + cross-sectional questionnaire study
Zohar and Feiz (2001)	children with more routines are at risk for OCD; normal routines are associated with more maladaptation and anxiety	X		routines and OC symptoms	228	community sample of children (2–5 years old)	cross-sectional questionnaire study
Bolton et al. (2002)	more magical thinking is associated with more OC symptoms	X		normal magical thinking and OC symptoms	127	community sample of children & adolescents (5–17 years old)	cross-sectional questionnaire study
Evans et al. (2004)	common underlying neurobiology	X		thoughts and obsessions; rituals and compulsions	/	children etc. with OCD + controls	review study
Santesso et al. (2006)	continuum at a psychophysiological level	X		rituals and compulsions	37	typically developing children (10 years old)	EEG study
Boyer and Lienard (2006)	continuum between normal, sub-clinical and clinical manifestations	X		thoughts and obsessions; rituals and compulsions	/	children etc.	review study
Pietrefesa and Evans (2007)	emotional, cognitive and neuropsychological continuities; fears-rituals relation increases with age and becomes a maladaptive coping	X		rituals and compulsions	42	typically developing children (4–8 years old)	neurological tasks + cross-sectional questionnaire study
Laing et al. (2009)	normative anxieties and rituals change in content over time as a function of altering developmental salience, in a similar fashion to OCD	X		normative anxieties and obsessions; rituals and compulsions	142	community sample of children & adolescents (7–16 years old)	semi-structured interview + cross-sectional questionnaire study
Bolton et al. (2009)	genetic evidence for continuum; children with more routines are at risk for OCD; more routines are associated with more OCD	X		routines and OC symptoms	4662	community sample of children (6 years old)	cross-sectional twin study
Evans and Maliken (2011)	continuum at a psychophysiological level	X		rituals and compulsions	12	typically developing children (6–12 years old)	EEG study
Glenn et al. (2012)	children with more routines are at risk for OCD; more routines are associated with more OCD and anxiety; routines decline with age but remain in a minority	X		routines and OC symptoms	1369	typically developing children (2–11 years old)	cross-sectional questionnaire study
Glenn and Nananidou (2016)	more routines are associated with more OCD and anxiety (cross-sectional); routines decline with age; anxiety and OCD incline	X		routines and OC symptoms	109	typically developing children (8–18 years old) (FU Glenn et al., 2012)	longitudinal questionnaire study
Zohar and Dahan (2016)	normal routines are a mild sub-clinical form of OCD and associated with executive functioning delays and anxiety	X		routines and OC symptoms	1345	community sample of children (2–6 years old)	cross-sectional questionnaire study

differences in timing, content and severity. The results revealed no differences in type or number of superstitions between the two groups, although parents of OCD children described significantly more rituals at a younger age. However, this research design was not conclusive as to whether this perception actually reflected the presence of early OCD manifestations or was merely an artefact of biased parental recall, because when early rituals that resembled OC symptoms ('pathological rituals') were excluded, the difference in number of 'normative rituals' between the groups disappeared. Hence, this design did not allow making firm conclusions about continuity.

From a *clinical/phenotypal* perspective, the first stream of evidence for continuity between normal rituals/routines and OC pathology can be traced back to Flament et al. (1988) and Swedo, Rapoport, Leonard, Lenane, and Cheslow (1989) who found that non-referred and OCD-diagnosed children and adolescents did not differ in intensity or number of rituals/superstitions. Bolton, Dearsley, Madronal-Luque, and Baron-Cohen (2002) focused on superstitions/magical thinking in community children and adolescents (5–17 years old), and reported a significant correlation between magical thinking and OC symptoms, as well as between magical thinking and anxiety. Further, Glenn, Cunningham, and Nananidou (2012) also found a significant correlation between routinized compulsive-like behavior and OC symptoms in 1369 typically developing children (2–11 years old) and reported a significant linear decline of normative rituals/routines with age. However, a minority continued to have higher levels of normative rituals/routines up to age 11. These children were more at risk for developing OCD and their routines were positively associated with anxiety. Glenn and Nananidou (2016) conducted a six-year follow-up study including 109 children (8–18 years old). Again, routines correlated strongly with both OCD (cross-sectionally) and anxiety, but unfortunately no longitudinal routine-OCD correlations were reported.

In Glenn et al. (2012; 2016), children at risk for OCD showed more routines, and these routines – or superstitions (Bolton et al., 2002) – were strongly associated with anxiety. Others also focused on anxiety when investigating the continuity between rituals and OCD. Evans, Gray, and Leckman (1999) for instance, examined the relation between compulsive-like ritualistic behaviors and normal fears and anxiety in 61 typically developing children (1–7 years old). They hypothesized that, if normative compulsive behavior in children is indeed continuous with OCD compulsions, normative routines would – just as OCD compulsions – be correlated with efforts to control anxiety or fear. This was exactly the case in the studies of Glenn et al. (2012, 2016) and Evans et al. (1999) in which parallel correlational patterns were found between "normative compulsions and fears" and "OCD-related compulsions and anxieties" – supporting the continuity hypothesis. In a similar vein, Laing, Fernyhough, Turner, and Freeston (2009) found that in children both normative compulsive behavior and OCD compulsions were correlated with efforts to control anxiety. Indeed, worry was strongly related to ritualistic behavior in 142 community children and adolescents (7–16 years old). Together, these studies indicate that rituals, both at clinical and non-clinical levels, can be seen as coping mechanisms to alleviate anxiety.

From a *developmental* viewpoint, Laing et al. (2009) showed that the frequency of normative rituals declines with age and that normative anxieties and rituals change in content over time as a function of altering developmental salience. This also happens in OC symptoms, which are known to change in content and severity over time (Rettew, Swedo, Leonard, Lenane, & Rapoport, 1992). This similarity in the developmental trajectories of normal and clinical OC manifestations reflects continuity. Further elaborating on this developmental perspective, Evans et al. (1999) reported that when children get older, the relationship between rituals and fears increases, but in addition, compulsive rituals in reaction to fears become less developmentally appropriate and thus resemble the OCD phenomenology more closely. This is congruent with others indicating that these early normative rituals have adaptive functions (e.g., reducing anxiety) and gradually decline as children mature, whereas a minority continues to have high levels of rituals and uses these as anxiety buffer when growing older (Glenn & Nananidou, 2016; Glenn et al., 2012; Laing et al., 2009; Pietrefesa & Evans, 2007). Relatedly, Zohar and Bruno (1997) found in 1083 community children and adolescents (8–14 years old) that ritualistic behavior is common and normative at young age, but then drops and even becomes rare and maladaptive at older ages (i.e., associated with anxiety). Nevertheless, even when ritualistic behavior is normative and adaptive at young age, children at the top of the distribution still can be shy, emotional and fearful, and could be more prone to develop OCD, as reported by Zohar and Felz (2001) studying 228 community children (2–5 years old). Zohar and Dahan (2016) concluded that highly ritualistic behavior is a mild sub-clinical form of OCD, reflecting continuity. More specifically, they showed in 1345 community children (2–6 years old) an inverse relationship between age and rituals, with highly ritualistic behavior being related to maladaptive OCD characteristics, such as fears, a negative-emotional temperament, emotional dysregulation and neurological deficits (e.g., executive function delays in shifting).

Other scholars investigated the continuity hypothesis from a *neurological* perspective. For instance, Evans, Lewis, and Iobst (2004) suggested in their review that there is continuity between the neurological correlates of OCD and the executive and neurobiological underpinnings of normative 'compulsive-like' behaviors in children. They argued that routines and their accompanying cognitive and emotional phenomena should not be seen as qualitatively distinct from pathological OC symptoms. Indeed, evidence indicates that these normal and pathological variants share a common underlying neurobiology in terms of impaired motor-suppression, response inhibition and set-shifting ability (Evans et al., 2004), as well as enhanced visuospatial abilities, as found in 51 typically developing children between 3 and 13 years old (Evans et al., 2001).

In another review, Boyer and Lienard (2006) referred to continuity between normal, sub-clinical, and clinical OC manifestations. They attempted to explain the presence of ritualized behavior in both normative and pathological circumstances, based upon a model unifying neuropsychology, neuroimaging and evolutionary anthropology evidence. This model refers to a 'precaution system' geared to the detection of, and reaction to, inferred threats. Normal activation of this system explains rituals in community participants, whereas the absence of negative feedback to the appraisal of potential threats – resulting in doubts and repetitive actions – can be observed in OCD.

Evidence for continuity was also found in EEG studies in typical children. Evans and Maliken (2011) reported that cortical activity of 12 children (6–12 years old) showed a linear association to rituals when exposed to visual asymmetry, suggesting that psychophysiological factors that are implicated in OCD also exist in normal rituals. In line with this, Santesso, Segalowitz, and Schmidt

(2006) found in 37 children of 10 years old that similar neural processes underlie both the pathological and normative aspects of repetitive behavior, supporting continuity at a psychophysiological level. Thus, EEG research supports the continuity between the observed brain-behavior links in OCD and normative variants of repetitive behavior (Evans & Maliken, 2011; Santesso et al., 2006).

Similar to Zohar and Dahan (2016), Pietrefesa and Evans (2007) studied ritualistic and compulsive-like behavior, anxiety and neuropsychological functioning in 42 typically developing children (4–8 years old) and showed that normal routines were associated with more anxiety and limited neurocognitive functions (response inhibition and set-shifting deficits) that also underlie OC symptoms. They concluded that there are emotional and neuropsychological continuities between normative and pathological ritualistic behavior. Hence, the well-supported links between rituals, anxiety and neurological functioning indicate that compulsive behaviors exhibited in normal development and OCD involve similar underlying factors (Pietrefesa & Evans, 2007).

Finally, only one *behavioral-genetic* study investigated the relation between normative routines and OC symptoms and the extent to which this relation is genetically mediated. Bolton et al. (2009) found in 4662 twin-pairs of 6 years old a significant correlation (0.40) between childhood routines and OC symptoms, which was completely attributable to genetic effects (i.e., the continuity is explained by shared genes). Although the cross-sectional nature of the study does not permit causal inferences, the results indicate that elevated levels of normative rituals/routines in childhood constitute a risk for developing OC symptoms. Hence, this groundbreaking study provided the first robust behavioral-genetic evidence for the continuity between normative rituals/routines and OC symptoms.

Development of an integrative conceptual model

Based on the review, we propose a model that (1) describes the ‘normative rituals/routines and OC symptoms’ relation, and (2) integrates factors that are important to systematically consider in evaluating the maladaptive character of the presented rituals/routines.

- (1) First, the results of the systematic review support the hypothesis that normative rituals/routines and OC symptoms in children and adolescents can be situated on a continuum that can be conceptually represented by a Continuity Model (Fig. 2/Part 1).
- (2) Second, by carefully structuring the results of the 18 included studies, it was clear that several studies exclusively focused on

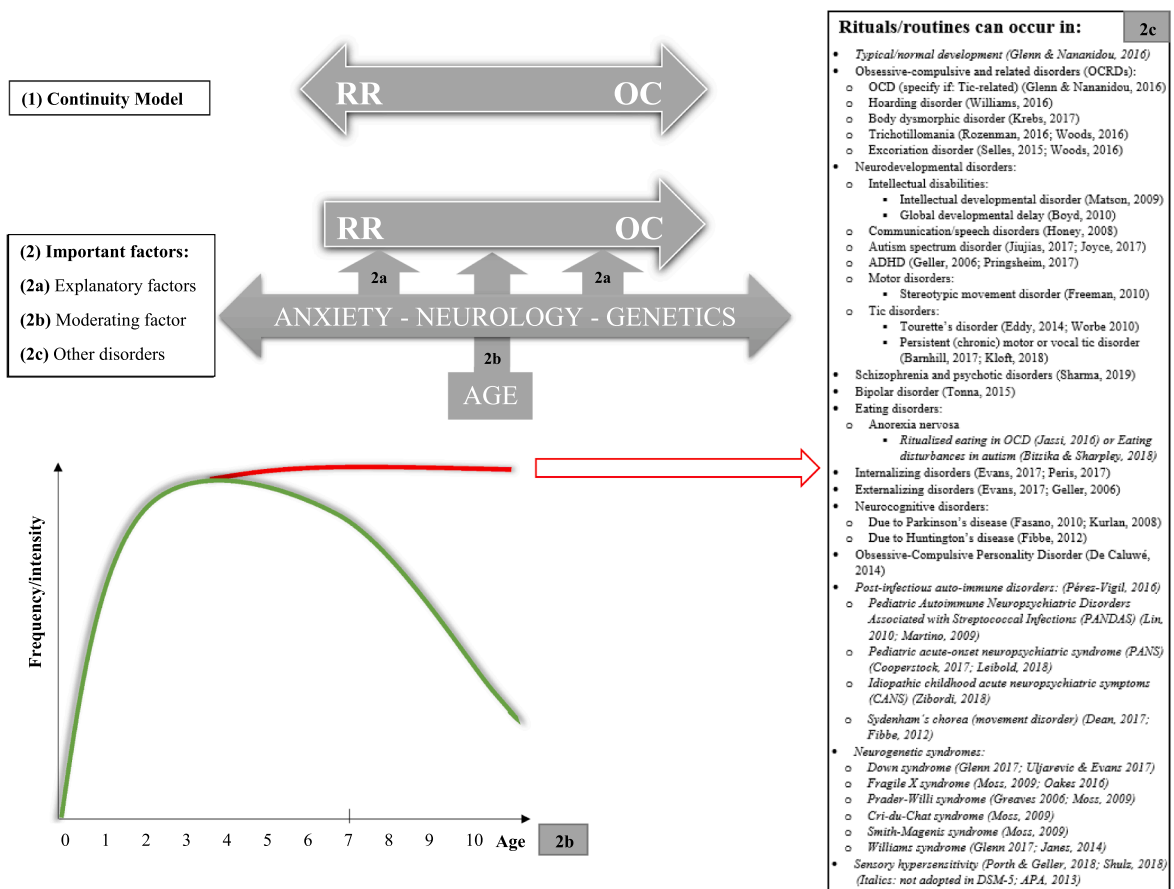


Fig. 2. Integrative conceptual model of the relation between normative rituals/routines (RR) and OC symptoms at a young age. (See above-mentioned references for further information.)

the association between normative rituals/routines and OC symptoms (Flament et al., 1988; Leonard et al., 1990; Swedo et al., 1989), whereas others also investigated additional important factors that can have a significant *explanatory/etiological* (anxiety, neurology, genetics; 2a) or *moderating role* (age; 2b).

(2a) More specifically, the results reveal important etiological factors at different levels of behavioral analysis, including the phenotypic (anxiety), neurobiological, and genetic level (Larsen & Buss, 2014). Hence, anxiety, neurology and genetics may *explain* the continuity between normative rituals/routines and OC symptoms, as they are underlying factors of both the normative and pathological variant of the continuum. Anxiety, for instance, is not only associated with both variants, but evidence also confirms that ‘anxiety reduction’ is one of the functions of both normative rituals/routines (adaptive coping at younger ages) and OC symptoms (increasingly maladaptive coping at older ages) (Evans et al., 1999, 2004; Glenn & Nananidou, 2016; Glenn et al., 2012; Laing et al., 2009; Pietrefesa & Evans, 2007; Zohar & Bruno, 1997; Zohar & Dahan, 2016; Zohar & Felz, 2001). Next to anxiety, both constructs are characterized by limited neurocognitive functions (motor-suppression, response inhibition and set-shifting) as well as enhanced visuospatial abilities (Boyer & Lienard, 2006; Evans et al., 2001, 2004; Pietrefesa & Evans, 2007; Zohar & Dahan, 2016) and similar EEG measured cortical activity (Evans & Maliken, 2011; Santesso et al., 2006). Behavioral-genetic research also indicates that the strong relation/continuity between normative rituals/routines and OC symptoms is completely explained by shared genes and that high levels of normative rituals/routines constitute a risk factor for developing OCD (Bolton et al., 2009). Other researchers (Glenn et al., 2012; Zohar & Dahan, 2016; Zohar & Felz, 2001) also suggested this latter point. Overall, evidence clearly indicates that normal rituals/routines and OCD involve similar underlying liability factors (anxiety, neurology, genetics), reflecting the emotional, neuropsychological and genetic continuities between normative and pathological ritualistic behavior. Together, these explanatory/etiological factors are integrated in the conceptual model (Fig. 2/Part 2a).

(2b) Based on the results, we further propose that age is a potential *moderator* as it reflects the position of ritualistic behavior on the normality-pathology continuum, with rituals/routines in younger children being normative (given the limited neurocognitive capacities) and even adaptive (e.g., as coping mechanism), but increasingly becoming rare and maladaptive with increasing age (e.g., reflecting OCD) (Evans et al., 1999; Glenn & Nananidou, 2016; Glenn et al., 2012; Laing et al., 2009; Pietrefesa & Evans, 2007; Zohar & Bruno, 1997). Thus, age is an important factor in evaluating whether rituals/routines are normal or maladaptive. In young children, there is a normative peak of rituals between the ages of 2–6 (Evans et al., 1997; Glenn et al., 2012). Around the age of 7 (Glenn & Nananidou, 2016; Glenn et al., 2012), rituals usually start to decrease as older children and adolescents do not need them anymore to alleviate anxiety, gain control over the environment and free up cognitive capacity. Instead, they have obtained more efficient coping mechanisms due to their development in executive functioning skills and maturation of the prefrontal cortex, usually starting at age 5 (Pietrefesa & Evans, 2007; Zohar & Bruno, 1997; Zohar & Dahan, 2016). In a minority, however, the amount and severity of rituals/routines stays high when growing older. These children continue to rely on this coping mechanism that becomes increasingly maladaptive and may develop into OCD. This moderating effect of age is reflected in the model and in a graph where the normative and pathological developmental trajectories of rituals are conceptually depicted, relying on different sources of evidence (Evans et al., 1997; Glenn & Nananidou, 2016; Glenn et al., 2012) (Fig. 2/Part 2b).

(2c) Whereas the previous components were based on the 18 studies, this last part on the *other disorders* relies on the entire initial screening results to ensure that all (DSM-5) disorders were included that may comprise rituals/routines (e.g., OCD spectrum disorders [Geller, 2006] and beyond). After evaluating whether presented rituals/routines are normative versus maladaptive, it is useful to examine whether the maladaptive rituals/routines can be understood in the context of OCD or can be seen as a clinical symptom of other disorders. This additional focus is crucial from a treatment perspective, as different disorders may require different interventions. To facilitate this process, we integrated in the model an overview of all disorders that can be characterized by rituals/routines, including the most relevant literature (Fig. 2/Part 2c).

Discussion

Given the phenomenological similarities in the rituals/routines present in normative development and OCD, as well as in other disorders, the early identification and differentiation between normality and pathology is complex. The current study aimed to address this challenge by improving our understanding of the relationship between normative childhood rituals/routines and OC symptoms in two ways. First, the continuity hypothesis was investigated by systematically reviewing all literature on this relation. Second, based on this review process, an integrative conceptual model was proposed that not only describes, but also aims to clarify this relation by integrating important influencing factors.

A critical consideration of the continuity hypothesis

Most studies in the systematic review rely on cross-sectional associations and comparisons. This is a crucial first step, offering initial evidence for the continuity hypothesis. A few studies, however, transcend this level of evidence, such as those of Bolton et al. (2009) providing the first robust behavioral-genetic evidence, as well as Pietrefesa and Evans (2007) and Zohar and Dahan (2016), offering a combination of emotional and neuropsychological evidence. Glenn and Nananidou (2016) also exceeded the cross-sectional level by conducting the first (and only) longitudinal study. Unfortunately, correlations between routines at baseline and OCD at follow-up were not conducted, neither other longitudinal analyses, which could have revealed important information on the developmental pathways of routines toward OCD.

Moving the field forward: guidelines for further testing the continuity hypothesis

To advance the current level of evidence, we propose several strategies that may guide researchers who aim to further test the continuity hypothesis. First, it is necessary to use more advanced research designs. It is recommended to expand the current cross-sectional designs towards (prospective) longitudinal designs, starting at a very young age. This will help us to understand the variability among developmental trajectories, and to define those pathways that develop into maladaptation. Further, we advise using large multi-informant samples (Durbin & Hicks, 2014) because child-parent inter-rater agreement for OC symptoms is modest (Storch, Rudy, Wu, Lewin, & Murphy, 2015). Second, more rigorously testing the continuity hypothesis requires cutting-edge statistical techniques, such as structural equation modeling (testing the correlated change between routines and OC symptoms across time) and item response theory analysis (testing whether both constructs reflect the same underlying trait, with OC symptoms situated at the maladaptive continuum end) (Tackett et al., 2013). Finally, it is important to use adequate measures, not only in research, but also during clinical assessment to facilitate early identification and differentiation. To assess normative rituals/routines, we recommend the Childhood Routines Inventory-Revised (Evans et al., 2017), an excellent dimensional measure that can be used in typical and clinical populations. To assess OC symptoms, we also advise a dimensional measure that takes into account the multi-dimensional OCD nature (Leckman, Rauch, & Mataix-Cols, 2007) and age-specificity (Butwicka & Gmitrowicz, 2010; Geller et al., 2001). We recommend the Children's Yale-Brown Obsessive Compulsive Scale Second Edition (CY-BOCS-II; Storch et al., 2019), a semi-structured interview, or the Youth Obsessive-Compulsive Symptoms Scale (YOCSS; De Caluwé & De Clercq, 2014), a self-report questionnaire. Both also assess impairment, which is crucial to differentiate between normality and psychopathology (Clark & Rhyno, 2005; Evans, 2000). Whereas the CY-BOCS is mainly used in patients, the YOCSS is also applicable in typical youth. This may facilitate future OCD research because it allows investigating analogue samples, i.e., non-clinical samples where the often-occurring subclinical OC symptoms are considered as equivalents of OC symptoms observed in OCD (Abramowitz et al., 2014; Berry & Laskey, 2012; Clark & Rhyno, 2005). Moreover, the use of such designs reflects an important research implication that results from our continuity findings.

An integrative conceptual model: implications for theory, practice and research

Based on the systematic review, an integrative conceptual model was proposed that represents the continuity between normative rituals/routines and OC symptoms at a young age, i.e., the Continuity Model. This offers theoretical implications on the nosology of OCD, which is completely in line with recent developments on the classification of psychopathology, such as the Hierarchical Taxonomy of Psychopathology (HiTOP) (Kotov et al., 2017; Krueger et al., 2018). More specifically, shortcomings of traditional approaches resulted in efforts to classify psychopathology quantitatively, offering extensive support for the continuity (versus discreteness) of psychopathology. This paradigm shift remains a work in progress but the current study adds to its development.

Our conceptual model also integrates important factors that may systematically affect this continuity. Based on this model, we can formulate several recommendations for clinical practice. During the early detection and differential diagnosis procedures, clinicians should note that higher anxiety and limited neurological functioning (motor-suppression, response inhibition and set-shifting) are not only characteristic of OCD but also of normative rituals/routines, as they are underlying *explanatory/etiological* factors of both constructs on the continuum, and therefore may have less differentiating value (Boyer & Lienard, 2006; Evans & Maliken, 2011; Evans et al., 1999, 2001, 2004; Glenn & Nananidou, 2016; Glenn et al., 2012; Laing et al., 2009; Pietrefesa & Evans, 2007; Santesso et al., 2006; Zohar & Bruno, 1997; Zohar & Dahan, 2016; Zohar & Felz, 2001).

Probably more helpful in distinguishing normal and pathological rituals is the child's age. Age has a *moderating* role because in younger children, rituals/routines are normal and even adaptive, but become increasingly rare and maladaptive with older age. So with maturation, most children show a linear decline in rituals. However, a minority that shows persistent higher levels of rituals across time (which were normative at younger ages) is at risk for developing OCD (reflecting treatment targets) (Evans et al., 1999; Glenn & Nananidou, 2016; Glenn et al., 2012; Laing et al., 2009; Pietrefesa & Evans, 2007; Zohar & Bruno, 1997). Research indicated that the age of seven might represent an important turning point (Glenn & Nananidou, 2016; Glenn et al., 2012) at which children naturally start to decline in ritualistic behavior as their adaptive function changes. However, we argue that this cut-off is highly idiographic and we need to take into account the child's maturation and mental age. Mental age is even more important than chronological age, as indicated for instance by research in children with Down syndrome, where elevated levels of rituals persist longer and are longer used to manage fear and unpredictability, relative to typical children of similar chronological ages (Evans, Canavera, Kleinpeter, Maccubbin, & Taga, 2005). Moreover, before treating rituals in children, it is always crucial to consider their developmental appropriateness and to reserve treatment for when impairment is present.

Early detection and differential diagnosis is also highly complicated by the fact that these rituals/routines do not only occur during normal childhood development and OCD, but also in *other disorders*. To facilitate this, we provided an overview of all disorders that may include rituals/routines, which the clinician might examine – reflecting suggestions for further clinical assessment. Moreover, given that the similarity between normative rituals/routines and OC symptoms is particularly present during early childhood development (Zohar & Bruno, 1997), we advise to timely and routinely screen children (Geller et al., 2012).

In addition to the recommendations for clinical practice, the integrative conceptual model also offers guidelines for further research. One important direction may be the study of *other explanatory factors* in addition to the currently addressed 'anxiety' at a phenotypical level. Besides anxiety, also 'not just right experiences' (NJREs) and disgust have been proposed as important emotion-related motivators underlying OCD (Berle & Phillips, 2006; Coles, Heimberg, Frost, & Steketee, 2005). However, they remain understudied so far, especially in youth. Some of the recent exceptions are studies of Cervin and colleagues (Cervin & Perrin, 2019;

Cervin, Perrin, Olsson, Claesdotter-Knutsson, & Lindvall, 2020) who showed that anxiety and NJREs are significantly correlated with OCD, though the association between disgust and OCD is less strong, just as in adults (Berle & Phillips, 2006; Berle et al., 2012; Sica et al., 2019). Future evidence in this regard may broaden the current integrative conceptual model towards other important explanatory factors.

A final suggestion for further research concerns the *other disorders* from the integrative conceptual model. In the current review, we investigated the continuity between normative rituals/routines and OCD specifically, but this can be replicated for all other disorders mentioned in the model (i.e., obsessive-compulsive and related disorders, autism spectrum disorders, etc.). We expect that for some disorders the same continuity model will hold (e.g., shared anxiety and neurobiological/genetic links), whereas for other disorders this may not be the case. For instance, anxiety is potentially an explanatory factor for the rituals/routines seen in some other obsessive-compulsive and related disorders, but not for those seen in some neurogenetic syndromes (e.g., Prader-Willi syndrome). Several studies already investigated this, and although these are very important, most of them exclusively focused on the phenotypical level, comparing for instance the level of repetitive behaviors in children with a typical development, children with OCD and children with autism spectrum disorders (Zandt, Prior, & Kyrios, 2007). Moreover, even if the focus is on explanatory factors (e.g., comparing neurocognitive functioning in children with OCD, hair pulling disorder and no disorder; Wilton et al., 2020), the effects are often moderate and contrasting, providing both evidence for and against the same continuum (Stein, 2000, 2019; Zandt, Prior, & Kyrios, 2009). Hence, the finding that rituals/routines belong to many disorders does not mean that the same continuity model might apply to all of them. Only future research can elucidate this, preferably by relying on a (meta-analytic) review study for each candidate disorder.

Conclusion

This study attempted to contribute to a better understanding of the relation between early OC symptoms and normative rituals/routines by critically reviewing the evidence for the continuity hypothesis. The systematic review results supported continuity, but several strategies concerning designs, statistics and measures can be suggested to stimulate further research. Moreover, an integrative conceptual model was proposed that addresses this continuity from an etiological perspective on different levels of behavioral analysis, also including moderating (age) factors of the extent to which routines/rituals are evaluated as maladaptive, as well as other disorders that can include rituals/routines. This model may offer various implications for theory, clinical practice and future research. It is hoped that progress in this field will result in an earlier, tailor-made treatment of OCD and its prevention.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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